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SHORT REPORT

Pancreaticoduodenal Artery Aneurysm Ruptured into Duodenum, Associated with Celiac Trunk Stenosis. Case Report and Review of Literature

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Pancreaticoduodenal artery (PDA) aneurysm associated with a celiac artery (CA) occlusion or stenosis is an uncommon event. We report the case of a 63-years old man who presented with acute abdominal pain radiating to the back. During the hospital stay, the patient had an episode of severe hematemesis. He had a gastroscopy and then a surgical exploration. However only with arteriography we found a PDA, which had ruptured into duodenum. The aneurysm was associated with a stenosis of the celiac trunk and was supplied by a dense network of collateral vessels from the SMA. The patient was successfully treated with embolization and was discharged on the 64th postoperative day. Short term and mid term follow-up was uneventful. This case shows the difficulty in diagnosing these rare events in time, indicating that angiography is indispensable to establish a diagnosis and enable nonsurgical treatment.

Keywords: Pancreaticoduodenal artery aneurysm; Celiac artery stenosis; Surgery; Embolization.

Introduction

Pancreaticoduodenal artery (PDA) aneurysm associated with celiac artery (CA) occlusion or stenosis is extremely rare, and occurs in 2% of all visceral aneurysms.[1] The number of reported cases has gradually increased due to recent advances in non-invasive diagnostic imaging modalities such as computed tomography (CT) and ultrasonography.

Aneurysms involving PDA, often develop spontaneously, however some authors suggest they are related to specific hemodynamic disorders. An early diagnosis before the occurrence of a fatal rupture is still difficult because of the rare incidence, nonspecific symptoms, and deep location in the body of such cases. We report herein the case of a PDA aneurysm ruptured into duodenum, associated with a CA stenosis in a patient successfully treated with embolization. Our case shows the difficulty in diagnosing these rare events in time, and in activating prompt treatment to stop bleeding.

We reviewed the literature of true ruptured and non-ruptured pancreaticoduodenal artery aneurysms associated with a celiac artery lesion.

Case Report

A 63-year old man (weight, 65 kg; height 166 cm), was admitted with acute abdominal pain radiating to the back and anorexia. There was no history of smoking or excessive alcohol consumption and there had been no episodes of pancreatitis, hypertension, portal hypertension, or abdominal injury. No signs of vasculitis or collagen disease could be found on examination. On admission, he was pale and in pain.

On examination he was apyrexial, his blood pressure was 110/70 mmHg and his pulse 84 beats/min. He had a markedly distended abdomen with pain in

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the right hypochondrium and weak bowel sounds, but no rebound tenderness or guarding. Blood tests revealed a haemoglobin of 10.1 g/dl, a white blood cell count of 11 900/mm,[3] and a serum albumin of 2.8 g/dl. The electrolytes, liver function parameters, and serum amylase levels were within the normal ranges. His abdominal pain increased and the patient had nausea and vomiting. A nasogastric tube was inserted and revealed active bleeding. After patient's hemodynamic stabilization, an operative gastroscopy revealed massive bleeding coming from the second part of the duodenum, but failed to stop the bleeding.

The patient therefore underwent an emergency laparotomy which revealed a duodenum filled with blood and involved in a fibrotic process. After kocherisation of the duodenum a duodenal ulcer-like lesion, sized 1,5 cm in maximal diameter was detected, apparently causing the bleeding. After duodenal suturing and accurate hemostasis, the bleeding stopped. However at the end of the operation, there was a reprise of bleeding. The patient therefore, underwent an emergency operative arteriography.

The arteriography revealed a pancreaticoduodenal artery aneurysm 10 mm in diameter ruptured into duodenum associated with a tight stenosis involving the celiac trunk and a supplied by a particularly dense network of collateral vessels of pancreaticoduodenal arcades and superior mesenteric artery (Fig. 1).

A selective catheterization and embolization of inferior-anterior pancreaticoduodenal artery and inferior-posterior pancreaticoduodenal artery was performed.

Catheterization and embolization of mesenteric-hepatic anastomotic arcades, permitted a complete thrombosis of the vessels supplying the aneurysm, while preserving the gastroduodenal artery (Fig. 2).

The postoperative course was complicated by a duodenal fistula, treated with conservative therapy. The patient was discharged on the 64th postoperative day. Short term and mid term follow-up was uneventful.

Discussion

Aneurysms of the pancreaticoduodenal artery are rare (2% of all visceral artery aneurysms) and nearly 30% of these aneurysms evolve as complication of acute or chronic pancreatitis. There seems to be no gender predilection and the most common age of involvement is the sixth decade. The association with celiac artery occlusion was first reported by Sutton and Lawton in 1973.[2] Since then, 57 cases have been reported.[3–15] Celiac artery occlusion or stenosis was found in 63% of all cases with PDA aneurysm,[8] and this was an incidental finding in about 40% of the cases where there was no rupture.

With the increasing efficiency of CT and angiography, many aneurysms are being discovered before rupture or thrombosis in patients being evaluated for abdominal pain or other disease. Despite these advances, a significant number of vascular aneurysms are not discovered until patients present with rupture.

Pancreaticoduodenal artery aneurysms often have symptoms of abdominal pain before rupture.[1] The difficulty to reach a correct diagnosis explains the high mortality rate with rupture (26% to 50%) and supports the need for early diagnosis and treatment.[1,5]

Our literature review, wants to point out the attention to true pancreaticoduodenal artery aneurysms

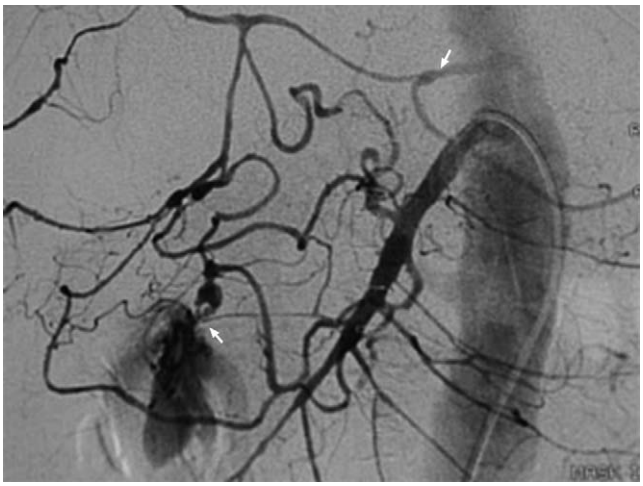

Fig. 1. Arteriography showing the celiac trunk stenosis and the pancreaticoduodenal artery aneurysm ruptured into duodenum.

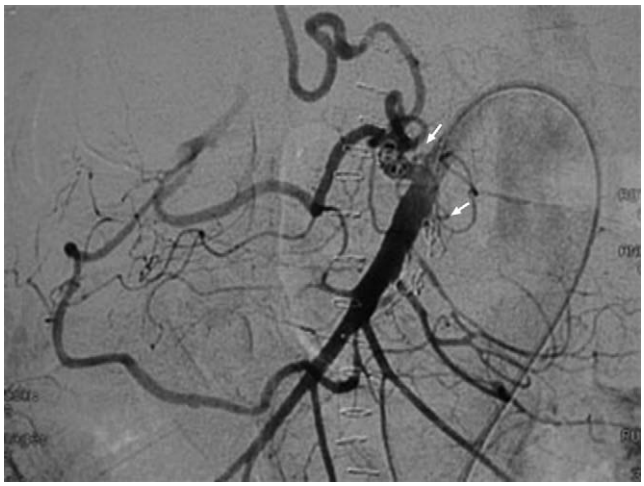

Fig. 2. Selective catheterization and embolization of the pancreaticoduodenal and superior mesenteric artery arcades supplying the aneurysm.

associated with celiac trunk stenosis, evaluating its correct management.

True aneurysms are often hard to differentiate from false aneurysms. Several findings suggested that our patient had a true aneurysm of the pancreaticoduodenal artery: the associated celiac trunk lesion, the absence of acute or chronic pancreatitis, and the dense arterial network connecting the superior mesenteric artery to the celiac artery.

False pancreaticoduodenal aneurysms, secondary to local injury, are mainly seen with acute or chronic pancreatitis.

True aneurysms are found in men and women equally and in patients with a wide age range: mean 60 years.[16,17] True pancreaticoduodenal artery aneurysms are especially rare and various causes have been reported including arteriosclerosis, infection, fibrodysplasia,[18] and mainly, stenosis or occlusion of the celiac trunk.

A celiac lesion in fact is acknowledged as a major cause of development of an aneurysm of the pancreaticoduodenal artery. Its prevalence varies from 63% to 74% in different series.[2,8,17] Sutton and Lawton[2] proposed that the increased blood flow in the peripancreatic arterial network provided collateral supply for revascularization of the celiac trunk, thereby dilating the vascular walls until an aneurysm developed.

Their hypothesis receives support from four reported cases that showed that simple revascularization of the celiac trunk, with surgical decompression through arcuate ligament section, or direct revascularization, led to complete aneurysm regression at angiographic followup.[19–21]

Our patient had a relatively small aneurysm in diameter. Most pancreaticoduodenal artery aneurysms range from 8 to 30 mm in diameter.[8]

Our review suggests that size is not a factor for rupture; all non-ruptured aneurysms measured more than 10 mm in diameter, whereas most ruptured aneurysms measured less than 10 mm. Most ruptured aneurysms manifest clinically with non-specific abdominal pain,[11,17,22–33] and in a few cases there is an acute abdominal syndrome associated with bleeding into the peritoneal cavity, and ultimately hemorrhagic collapse.

These aneurysms usually rupture into the retroperitoneal space around the pancreas, but also in the peritoneal cavity,[28,29] or, exceptionally, into the digestive tract, the duodenum,[26] or the Wirsung canal.[31] In our patient, abdominal ultrasonography could not identify any relevant finding because of the presence of intestinal gas. Moreover a CT scan was deemed unsafe because of hemodynamic instability. The first diagnostic step to identify the exact site of bleeding was a gastroscopy because the clinical presentation simulated a peptic ulcer. The surgical operation did not identify the presence of a visceral artery aneurysm, because of its appearance with an ulcer-like duodenal lesion. Only arteriography, identified the presence of a visceral artery aneurysm and showed an associated lesion of the celiac trunk. Our case report emphasizes therefore the role of angiography in localising the source of bleeding and its treatment by embolization.

The literature review showed that the current management of pancreaticoduodenal artery aneurysm depends on the presentation of the patient, comorbidities and risk factors. It consists of no treatment, surgery, embolization therapy, and treatment of the celiac trunk lesion. No treatment is an exceptional option, reported only twice in 39 case for non-ruptured aneurysms: in one case the patient refused therapy; in the second case there was life-threatening risk in a patient with cirrhosis, who died within a few days, of hepatic insufficiency reports.[2,17] Surgery was most often used.

The surgical procedure involved resection or simple exclusion of the aneurysm from the circulation. Surgical management through overseeing the aneurysm and ligating the feeding arteries, associated with decompression by section of the large left pillar of the arcuate ligament, may be hazardous, because of the high risk of rupture owing the presence of multiple pancreatic vessels that communicate with the aneurysm.[8] Therefore transcatheter embolization is the only alternative treatment in such cases.[16,34]

Conclusion

Aneurysm of the pancreaticoduodenal artery associated with celiac trunk stenosis is an exceptional event, but requires a prompt management because of its risk of rupture regardless of size. Rupture is often the initial symptom so, intraoperative isolation and control, can be difficult and hazardous. Our patient had no risk factors or symptoms that could justify a visceral artery aneurysm rupture, so he was treated for a gastro-intestinal bleeding.

Otherwise, in this case, the right management would have been not to perform surgery after endoscopy failed to stop the bleeding, but a minimally invasive treatment through angiography.

Arteriography can establish a diagnosis, and enable non-surgical treatment with embolization.

However in cases of a stenosis of the celiac trunk, caused by median arcuate ligament compression, an elective surgical decompression should be considered, to prevent the risk for recurrent aneurysm.

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